

Streeter dysplasia, from pelvic to digits: a case report

by Komang Agung Irianto

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Streeter dysplasia, from pelvic to digits: a case report



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Komang Agung Irianto S,^{1,2*} Luh Gede Djatu Anggita Dewi,² Gana Adyaksa¹

ABSTRACT

Background: Streeter dysplasia is a term to describe fetal congenital syndrome which mainly characterized by constriction band on appendages, prenatal amputations of extremities, and acrosyndactyly. This syndrome has wide range of clinical manifestation between patients, as reflected by many other terms to describe this syndrome.

Case: The author reported five cases of Streeter dysplasia with constriction band on different locations of the body, with a patient

having a constriction band around pelvic and other multiple anomalies, patient with constriction around leg and caused acute limb ischemic, and several cases of acrosyndactyly around hand and foot.

Result and Conclusion: Constriction band release surgery, as well as correction surgery for other abnormality was performed, either by direct closure or Z-plasty with satisfactory result in functional and aesthetic.

Keywords: Streeter dysplasia, constriction band, pelvic, limb ischemic, acrosyndactyly.

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¹Orthopedic and Traumatology Department, Airlangga University/ Dr. Soetomo General Hospital, Surabaya-Indonesia

²Surabaya Orthopedic and Traumatology Hospital

INTRODUCTION

Streeter dysplasia is a term that describes a congenital fetal syndrome which is mainly characterized by constriction ring on the limb, prenatal amputation of extremities and acrosyndactyly. There are a wide variety of other terms describing this syndrome, such as constriction band syndrome, annular band syndrome, annular constrictive rings, pseudoainhum, amniotic band sequence, amniotic band syndrome (ABS), and congenital constriction ring syndrome (CCRS).¹

In 1832, Montgomery introduced the term amniotic band constriction, which is a term that describes a congenital disorder with clinical manifestations including annular constriction in the extremities, oligodactyly, cleft lip and cleft palate, club foot, acrosyndactyly, and hemangioma. Other clinical manifestations including the absence of limb, short umbilical cord, craniofacial disorders, neural tube defects, and body wall defects such as gastroschisis.²

The prevalence of this syndrome is approximately between 1:1200 live births to 1:1500

live births. The prevalence of male than female was 0.91:1.44. This defect is more common in African-Americans compared to Caucasians. History of the disorder in the family are also very rare.³

Definite etiology of this syndrome is unknown, but it was believed to be caused from prenatal factors and emerged as a result of excessive contraction of the uterus muscles and bleeding from a marginal blood sinus.⁴ In 1930, George Streeter introduced intrinsic germ plasm defect theory that occur on the plates of embryonic and amniotic cavity. Streeter found a band of macerated epidermal layer and

other residues from local tissue defect.⁵ And in 1965 Torpin and Faulkner introduced extrinsic theory, the hypothesis that the separation of amnion from chorion during early pregnancy produces a tissue that floated free and then covered parts of the embryo as a ribbon or got ingested by the fetus. The band will hamper growth or causing structural defects in the fetus. The limb of the fetus that covered by the band may undergo vascular compression and necrosis. And oligohydramnios conditions will worsen the compression and deformity.⁶

The clinical spectrum of this syndrome is very broad, which would require a special approach for each case, either for diagnosis, treatment, and rehabilitation. In this case report, there are 5 cases of Streeter disease with clinical manifestations of constriction bands in different locations; one patient with a constriction band in the pelvis with multiple anomalies, one patient with constriction bands on leg causing acute limb ischemia, and three patients with constriction bands and acrosyndactyly on the fingers and toes.

CASE REPORTS

Case 1

An 18-month-old girl, was first brought to Surabaya Orthopedic and Traumatology Hospital at the age of 4 months, with a chief complaint of congenital abnormalities, who had a constriction band in the pelvis and the left inguinal region (Figure 1). The patient is the first child of 29-year-old mother and 32-year-old father, with no abnormalities found during antenatal, term pregnancy, and delivered

*Correspondence to: Komang Agung Irianto, S Orthopedic and Traumatology Department, Airlangga University/Dr. Soetomo General Hospital
komang168@yahoo.com

by sectio caesarea because of breech presentation. She also had right club foot, right knee extension contracture, bilateral DDH, occult spina bifida of L4-L5-S1, congenital thoracolumbar scoliosis, and flexion contracture of the right hip.

Constriction band in the region of the pelvis and groin were deep enough, but did not cause any pain and disruption on the distal neurovascular. By using ultrasound, it was found that the constriction band in the pelvis region has 17.3 mm of depth (Figure 2: a, b). Meanwhile, on the side of the left inguinal, constriction band was found in 13.6 mm on the lateral femoral AVN. Hip ultrasound showed 36.4° of α Angle on right hip and 54.6° of α angle on left hip. (Figure 2 c, d)

The patient underwent bilateral DDH correction followed by spica cast, and serial plastering to correct the club foot (Figure 3). Correction of the constriction band were performed in two stages. The first stage was the release of the anterior side



Figure 1 Clinical and radiological images of patient. There were constriction band in the pelvis and the left inguinal region

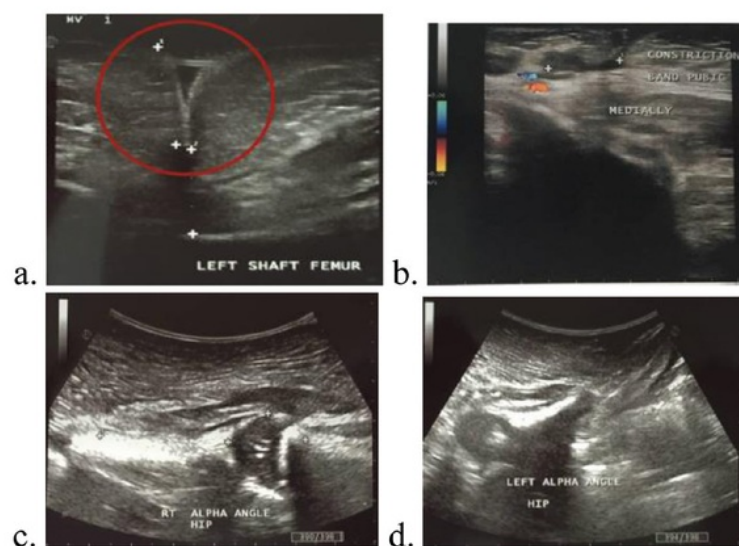


Figure 2 Ultrasound region of the pelvis and hip. a. The depth of constriction band region of the pelvis was 17.3 mm. b. Constriction band is 13.6 mm lateral to femoral AVN. Angle α hip sonogram (c. Right: 36.4° , d. Left: 54.6°)



Figure 3 a. Correction of bilateral DDH (pre-corrections, post-correction, and followed by spica cast). b. Serial plastering for the correction of club foot, followed by the application of Denis Browne splint

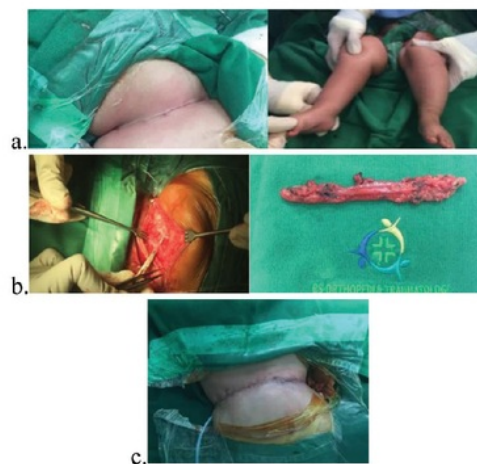


Figure 4 Constriction band release surgery. a. Step one, released the anterior side of constriction band, simultaneously with Achilles tenotomy. b. Step two, released posterior constriction band and excised fibrotic scar tissue. c. Closed the wound using direct closure technique



Figure 5 Post release constriction band. Results of post release anterior side is quite satisfactory, and the child is able to stand on her feet

of constriction band, which performed simultaneously with percutaneous Achilles tenotomy of right leg (Figure 4a). The next stage was the release of the constriction band on the posterior side. Constriction scar successfully removed and the wound closed with direct closure technique. (Figure 4b, c).

Eight months after surgery, the result was quite satisfactory. In addition, the child also has been able to stand on her own feet (Figure 5).

Case 2

A 9-month-old girl with a history of congenital constriction band on the left leg was brought to



Figure 6 Clinical pictures of the patient. Constriction band seemed located quite deep in the left crus, with the distal portion looks edema, shiny skin and cyanotic



a.



b.

Figure 7 Constriction band release surgery. a. The constriction band and vascular structure underneath was released. b. The defect was closed using Z-plasty



Figure 8 Clinical photos 8 months post-surgery. No constriction bands on the anterior, medial and lateral side of left leg

the Dr. Soetomo Hospital emergency department with chief complaints of swelling, pain and bluish on the child's left leg, which happened from a day before the admission (Figure 6). The constriction skin on the left leg was obtained from birth. There was no history of pain, swollen, and bluish leg before. The patient was the first child of the 26-years-old mother and 28-years-old father. Antenatal care was carried out by a midwife, and there was no abnormality during pregnancy. There were no history of illness or consumption of drugs or herbs. Patient were delivered spontaneously by the help of a midwife. Patient was diagnosed as having Streeter disease with acute limb ischemic.

Urgent release of constriction band was performed in this patient. Operations was carried out in one stage, the constriction band and vascular structures underneath was released. The defect was closed by using Z-plasty (Figure 7).

Eight months after surgery, the patient never had any complaint about swelling, pain and bluish on the left leg anymore. The scar looks good, and the patient was able to walk normally with his legs (Figure 8).

Case 3

A 9-month-old girl was taken to Dr. Soetomo pediatric orthopedic clinic hospital with complaints of fingers and toes that stuck and huddled, and there was a constriction in the skin since birth. The patient is the first child of the 27-year-old mother and 29-year-old father. There were no abnormalities during antenatal, delivered at term spontaneously with the help of a doctor. There were constriction band on thumb of right hand and acrosyndactyly of finger 3, 4, 5 of the right and left hand and also constriction band on finger 4 and 5 of the left and acrosyndactyly of finger 2, 3, 4 of the left foot (Figure 9).

Release of constriction band and acrosyndactyly of right and left hand, and left foot was performed in single stage. And finally, it was closed by using Z-plasty (Figure 10 and 11).

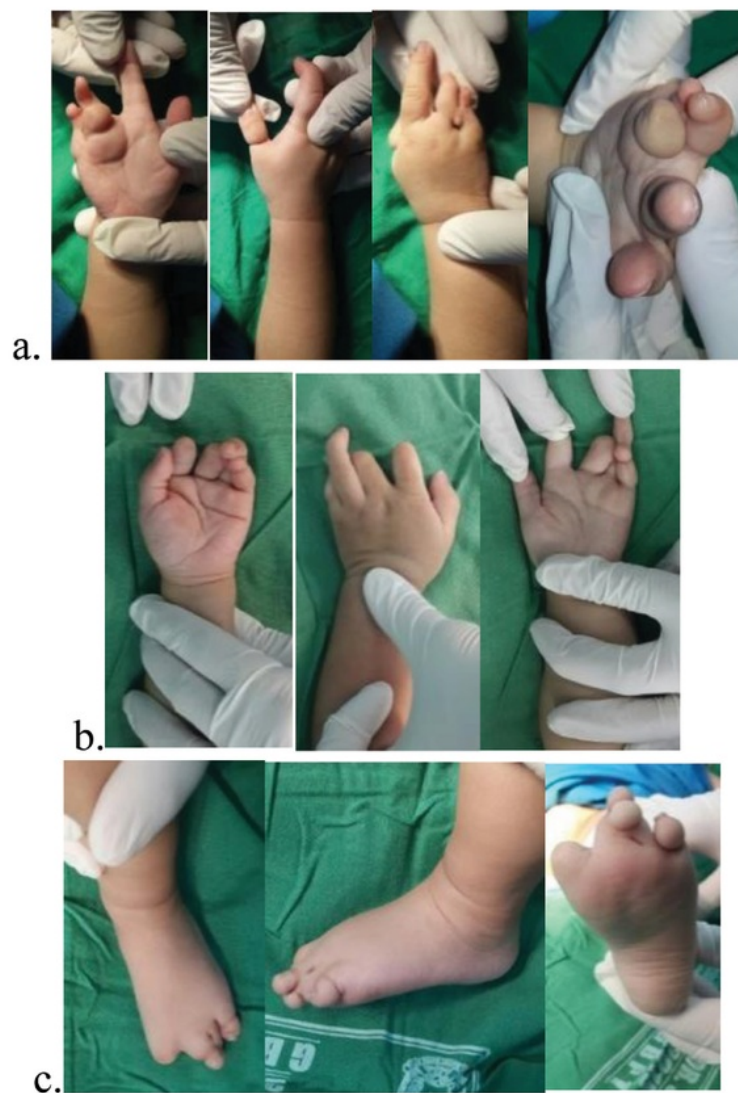


Figure 9 Clinical photos of patient. a. Constriction band on thumb and acrosyndactyly of finger 3, 4, and 5 of right hand. b. Acrosyndactyly of finger 3, 4, and 5 of left hand. c. Constriction bands on finger 4, 5 and acrosyndactyly of finger 2, 3, and 4 of left foot

Case 4

A 4-month-old boy was first brought to the Dr. Soetomo Orthopedic Clinic with chief complaint that there were skin indentations on the left leg and atrophy of the fingers of the left hand. The patient is the third child of 32-year-old mother and 33-year-old father. There was a history of taking medication to induce menstruation at the age of 8 weeks' pregnancy. The child was born at term spontaneously with the help of a midwife. The patient was diagnosed with constriction band of left leg and syndactyly of finger 2, 3, and 4 of the left hand (Figure 12).



Figure 10 Post release constriction band and acrosyndactyly. a. Right hand. b. Left hand c. Left foot



Figure 11 Clinical photo 7 days' post-surgery



Figure 12 Clinical Photos. a. Constriction band in left ankle. b. Syndactyly of finger 2, 3, 4 of the left hand

Case 5

An 8-year-old boy was referred to RS Dr. Soetomo Surabaya pediatric orthopedic clinic with complaint there were bondage under the skin on the left leg and the fingers of the right hand, and left foot fingers were attached.



Figure 13 One year after release constriction of the left ankle band and release syndactyly of finger 2, 3, and 4

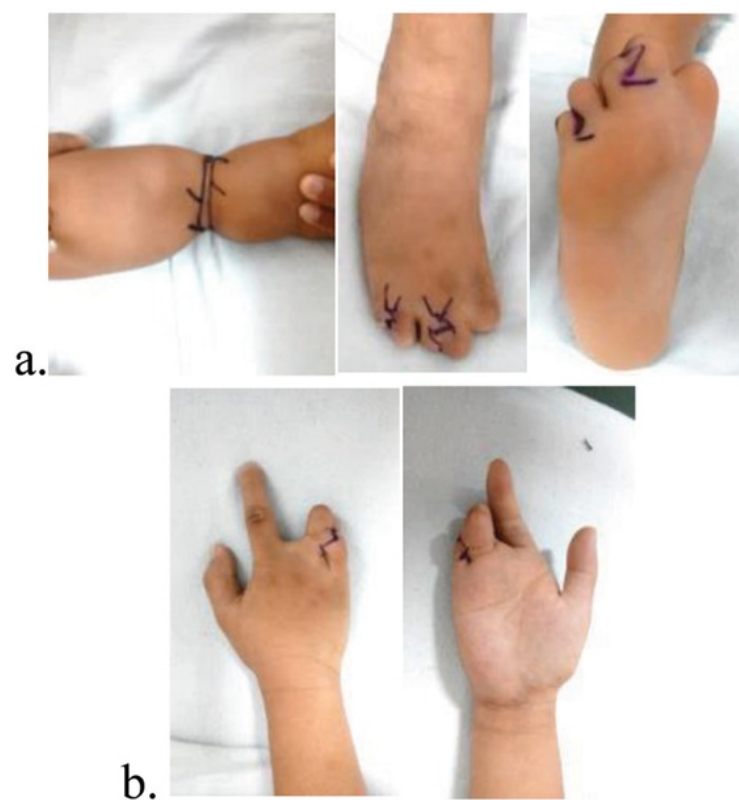


Figure 14 Clinical photos patient. a. Constriction band at left leg and syndactyly of finger 2, 3, 4, and 5 of right foot. b. Syndactyly of finger 3, 4, and 5 of left hand

Constriction band release was performed in the region of the ankle and fingers of the left hand by using Z-plasty (Figure 13).

The patient was the first child of 24-year-old mother and 26-year-old father. There were no abnormalities found during antenatal. The boy was born at term spontaneously with the help of a midwife. The patient had been able to walk since he was 15 months old. Patients often complain of pain in his left foot, but never got his left leg swollen or bluish. Patient was diagnosed with constriction



Figure 15 Clinical post-operation photos. a. Post release constriction of left leg by Z-plasty. b. Post release syndactyly of finger 2, 3, 4, and 5 of right foot, c. Post release syndactyly of finger 3, 4, and 5 of right hand



Figure 16 Clinical photos 6 months after constriction band release surgery of the left cruris and syndactyly release of the right hand and the foot

band region of the left cruris, syndactyly of finger 2, 3, 4, and 5 of right foot and syndactyly of finger 3, 4, and 5 of right hand (Figure 14).

Constriction band release of left leg, and syndactyly release of right hand fingers and foot was performed using Z-plasty (Figure 15).

Six months after surgery, post-operative wound of constriction band release on the left leg looked pretty fair. Patient also had never felt pain, swelling or bluish in his left foot anymore (Figure 16).

DISCUSSION

Congenital constriction band is defined as congenital abnormalities found in infants, in which the constriction ring depresses the soft tissue, ensnaring fingers and limbs.

In some rare cases, it can be found in the neck, chest, and abdomen. A coincidence from the medical history of the family is very rare, and genetic predisposition or gender differences factor were unknown. The pathogenesis factor was also not yet known.⁷

Although constriction band syndrome in the fingers and/or legs was widely reported and discussed, constriction band in the region of the trunk is very rare. Up until 2014, there were only five reports on constriction band syndrome in the trunk, with the location of the band at the umbilicus or above the umbilicus.^{8,9} Constriction band syndrome in the pelvic region, with a band located below the umbilicus, only had 6 patients reported, with 3 patients treated using Z-plasty and the remaining 2 patients did not receive surgical treatment.¹⁰⁻¹⁴

In the third, fourth, and fifth case, the main manifestation of the Streeter disease is a constriction band and acrosyndactyly on the hand and foot fingers. Although there is no widely agreed classification, clinical manifestations of these patients can be classified as Type 3 Patterson classification,¹⁵ that the primary manifestation is acrosyndactyly, or fenestrated syndactyly, which is a fusion of the distal cutaneous digit separation on the proximal side.

Proper time management needs to be applied in treating patients with Streeter disease. Congenital constriction band should be treated as soon as possible, in order to prevent further complication such as progression of the constriction or compression of nearby tissues. There were some cases of complaints of pain in amniotic constriction band located in pelvis when the patient gets older.¹¹ As in the second case, constriction bands on legs compressed the vascular and resulted in acute limb ischemic on the distal of the constriction. Therefore, urgent release of the constriction band was performed to prevent further complications.

The treatment of the constriction band, as well as other abnormality (for example bilateral DDH and club foot in the first case) were performed as early as possible, in order to prevent complications, improve functional and to pursue or optimize the growth of the patient, as well as to achieve a better aesthetic. In the first two cases, after the constriction band release operation was performed, the patients were able to stand and walk on two feet.

In amniotic band syndrome, imaging studies are essential to obtain accurate assessment of some important aspects of the constriction ring, such as the location of the ring, the depth, and important tissues or organs surrounding it. In the first case, the imaging of constriction band is very important because constriction ring is located in the pelvic

region, where there are many important structures in the pelvis and inguinal regions, particularly AVN. Therefore, ultrasound was chosen as imaging modality, since it is non-invasive, non-radiating and can help determine the depth of the constriction ring surrounding critical structures. In addition, an ultrasound examination in this case was also essential in assessment of stability of the hip by α -angle measurements.

One important thing in the surgical treatment of constriction band is to excise entire constriction scar or tissue fibrosis completely. Otherwise, the incidence of recurrence is high.¹⁶ A number of techniques have been reported to be capable to overcome this defect. Among them, the most widely used is the Z-plasty or W-plasty, with the development of two or three stages of operation to a single stage operation. However, one of the problems that often arise from these techniques was the healing of surgical wounds with the trend of local scar contracture and saw tooth disfigurement.¹⁷ The closure of the defect can also be done by using direct closure, as is done by Choulakian, in which he argued that in the constriction band excision it is not necessary to cover the shortfall or the sand glass deformity caused by Z-plasty, nor by the surrounding fat tissue debulking. By a simple excision and direct closure, eventually, the fat tissue will fill and seal the defect naturally.¹⁸ On the first patient, the size and depth of the network constriction ring has been predicted accurately by using ultrasound. The surgery was carried out in 2 stages, in which the fibrotic scar tissue was able to be excised completely, and then direct closure was performed. Closure by direct closure (without Z-plasty) proved to be effective and did not cause any problems later on, as reported by Rosson, where the post-operative wound by direct closure of constriction band region of the pelvis seemed natural and attractive.¹¹

Club foot is a disorder that commonly accompanies the constriction band syndrome. The incidence of club foot deformity in amniotic band syndrome is ranged between 12-58%, with 30% of it is bilateral. Two types of known club foot are rigid and paralytic.¹⁶ Gomez reported that 80% of neonates born with amniotic band syndrome and club foot has an annular constriction ring congenital at the ipsilateral limb.¹⁹ Often, compression band on peroneal nerve will give rise to distal neuropathy and result in paralytic club foot, due to eversion muscles paralysis. Furthermore, club foot in amniotic band syndrome responds to casting poorly. Gomez mentioned that club foot in amniotic band syndrome is very rigid and difficult to be corrected, and often require surgical treatment.¹⁹ In the first case, there is a club foot on the right leg without annular constriction band on the right leg. Club

foot in these patients is quite rigid, and treated with serial plastering. Achilles tenotomy was performed as well as the use of Denis Browne splint after serial plastering. This combination therapy has shown good results, in which eight months' post-surgery, the patient can stand on her own feet.

Apart of constriction band in the region of the pelvis and groin, Streeter dysplasia in the first patient in this report was accompanied by a wide range of other abnormalities such as club foot, bilateral DDH, scoliosis, spina bifida, and extension contracture of the knee. As already mentioned in the literature, the exact pathogenesis of this disorder patients was also not known yet. There was no abnormality in the family's medical history, and also there were no antenatal abnormality. Only positive result of CMV IgG found by laboratory examination (the first case), where there is no association found from the CMV infection and Streeter dysplasia. Most abnormalities in these patients as constriction band, club foot, bilateral DDH, scoliosis, and acrosyndactyly probably caused by the crush of the amnion or the wall of the uterus, according to the extrinsic theory. However, extrinsic theory is unable to explain the occult spina bifida from the first patient, where this disorder may be caused by a germ defects (neural tube defects), as described by Streeter by his intrinsic theory.

SUMMARY

Five cases of Streeter dysplasia, with constriction band in various locations, as well as other congenital abnormalities has presented, representing the wide range of clinical manifestations of this syndrome. Constriction band release surgery, either by using direct closure or Z-plasty, as well as correction for other accompanying abnormalities were successfully performed. Appropriate time and clinical management were optimally performed, in order to prevent complication, optimize function and developmental, and also to get better aesthetic.

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